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Shave Excision as an Adjunct to the Therapy of a Rhinophyma-like Complication in Post-kala-azar Dermal Leishmaniasis

Sir,

Post-kala-azar dermal leishmaniasis (PKDL) is an uncommon sequel in patients with a previous history of kala-azar (KA). We describe here a hitherto unreported mode of surgical excision for an unusual complication that did not regress following successful antimonial therapy.

CASE REPORT

A 40-year-old man from the eastern part of India presented with eruptions of 15 years' duration. They had commenced on the nose and cheeks, initially as transient erythema, leading to persistent induration and papules. Later, lesions appeared on the trunk, external genitalia and extremities, in that order. After a few years lesions had appeared on the tongue and glans penis. Five years prior to the onset of the eruptions he had been treated for KA elsewhere. Six years ago he had been diagnosed with PKDL and been prescribed injections of sodium antimony gluconate, but had never completed the course. The condition had then progressed and disfigured his face, which led to him presenting to us.

On examination erythematous induration was prominent on the face, studded with papules and nodules on the eyebrows, malar area, lips, chin and a few on the ears. On the nose they had coalesced, forming a large irregular nodule (Fig. 1). Well-defined, irregular hypopigmented macules were present on the back and trunk. Scattered papular lesions were seen in the axillae, trunk, upper limbs, penis and thighs. Nodular plaques were seen on the dorsa of the hands and scrotum. Papules were seen on the tongue and glans penis. The palms and soles appeared normal.

Systemic examination revealed no abnormality. Routine haemogram, urinalysis, and liver and renal function tests showed no abnormality. ECG and chest skiagram were within normal limits. Slit-skin smears stained with Giemsa and Ziehl-Neelsen technique revealed no organisms. Skin biopsy from the nose was taken for histopathology and culture. The former revealed a diffuse infiltration of the dermis by a dense mixed inflammatory infiltrate of lymphocytes, histiocytes and numerous plasma cells. Leishman-Donovan bodies (LDB) were identified in a few histiocytes. The epidermis showed hyperkeratosis and pronounced follicular plugging. Culture in Medium 199 containing 10% foetal calf serum, penicillin 10 u/ml, streptomycin 10 µg/ml and Hepes buffer pH 7.4 grew spindle-shaped, flagellated promastigote form of the parasites after 8 days of incubation at 24°C.

The patient was treated with 10 ml (1 g) of sodium antimony gluconate daily i.m. The nodules showed considerable regression after 6 weeks. Monthly ECG was done to keep a watch on cardiac function. After completing a total of 120 g in 125 days, he was asked to stop therapy. The indurated sites had completely regressed. The papules and nodules had subsided well in all areas except over the nose where the lesion had shrunk but still remained prominent. On the advice that the hypochromic macules and the nasal lesion would eventually disappear, he was kept under follow-up. Three months later he returned, stating that the lesion on the nose hampered social interactions (Fig. 2). Re-examination of the rest of the skin showed signs of subsided disease. The desire was to achieve a cosmetically acceptable result that was not socially disabling. We chose shave excision to clear the lesion on the nose. This was performed using a no. 11 scalpel blade under local anaesthesia. The bleeding points were controlled with pressure and light electrodesiccation. After completion, firm bandage was applied and changed every fourth day. Prophylactic broad-spectrum oral antibiotics were given for a week. The lesions healed without scarring in 10 – 14 days without any disfigurement of the nose

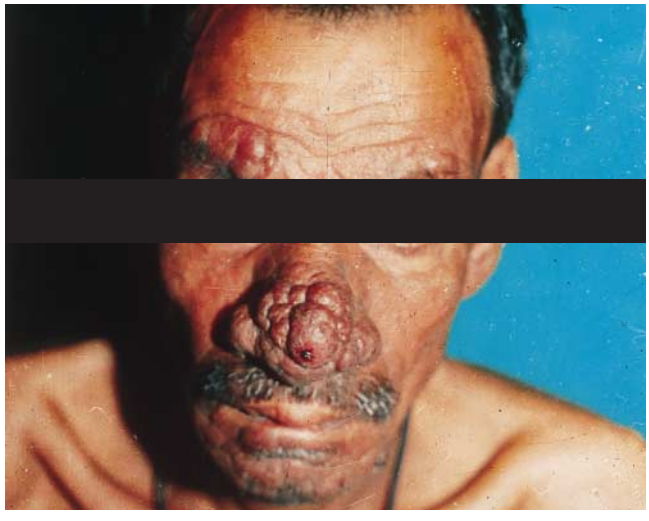

Fig. 1. Large irregular nasal nodule.

Fig. 2. Nasal lesion remnant on follow-up.

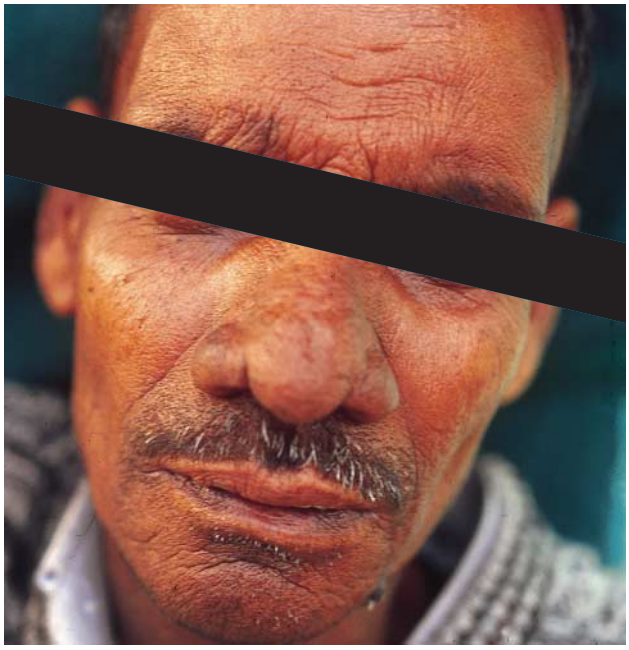

Fig. 3. Normal nose after surgery.

(Fig. 3). Histopathology of the shaved specimen showed multiple keratin containing cavities lined with squamous epithelium. Many pilosebaceous units were seen opening into each cavity. The inflammatory infiltrate in the surrounding dermis was sparse and no LDB were seen. These features were similar to that seen in rhinophyma.

DISCUSSION

Although the scourge of KA has been controlled with the advent of pentavalent antimonials, PKDL proves to be a refractory sequel. The lesions in PKDL nearly always commence on the face, attributed to photosensitivity, and later spread to the other parts of the body affecting at times the mucous membranes. They cluster in the central facial part including the forehead, nose, perioral area and the chin forming coalescing nodules that persist for years but rarely ulcerate. In advanced cases the lesions on the nose may spread to the anterior nares, but usually stop short of the mucosa. Only recently prolonged therapy with sodium antimony gluconate in a dose of 20 mg/kg i.m. daily for 120 days, sometimes longer or occasionally shorter, has been shown to be effective in PKDL (1, 2).

Diagnosis in our patient was based on demonstration of LDB in histopathology and isolation in culture. The serum of this patient had also revealed antibodies to the major antigens of Leishmania donovani in a recent study (3). Experience has shown that the papulonodular lesions in PKDL are the first to respond to antimonial therapy (4) and normal colour in the hypochromic macules appears gradually following completion of therapy (2). Barring the nasal lesion all the other plaques and nodules had completely regressed in our patient following regular treatment. So the possibility of drug resistant organisms or inadequate therapy did not arise. It may be because the nose is at times the site of severe involvement and takes more time to resume normal appearance. However the shrunken nasal lesion in this report appeared keratotic and circumscribed resembling a cutaneous horn. As it posed a social problem the final decision to perform shave excision was taken after 3 months of follow-up. The keratin-filled cavities seen on histopathology in the excised specimen are attributed to the extreme follicular plugging reported at times to give rise to epidermal cysts in PKDL (5). The nose being rich in pilosebaceous units and a favoured site in PKDL, development of a rhinophyma-like lesion may be an unusual complication for which surgical excision is recommended after completion of chemotherapy. Chronic inflammation and photosensitivity that accompany the dermatosis are likely to be the main factors contributing to this complication.

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